

## Obstructive gastric adenocarcinoma arising from heterotopic pancreas

Gastric cancer arising from heterotopic pancreas

Orhan Kalaycı<sup>1</sup>, Murat Özgür Kılıç<sup>1</sup>, Affan Çakır<sup>2</sup>, Zeliha Burcu Sarı<sup>3</sup><sup>1</sup>Department of Surgical Oncology<sup>2</sup>Department of Gastrointestinal Surgery<sup>3</sup>Department of Pathology, Eskişehir City Hospital, Eskişehir, Turkey**Abstract**

The stomach is the most common location of the heterotopic pancreas (HP). Most gastric lesions have benign behavior with mild or nonspecific symptoms. The limited number of malignant HP cases presenting with gastric outlet obstruction have been published. A 69-year-old man presented with epigastric distension, nausea, vomiting, and weight loss for two months. Upper gastrointestinal endoscopy revealed a dilated stomach and pyloric submucosal wall thickening causing complete outlet obstruction. Computed tomography demonstrated dilated gastric lumen with circumferential wall thickening in the pylorus. At laparotomy, the stomach was dilated, and a firm mass obstructing the lumen was palpated in the pyloric region. Subtotal gastrectomy with gastrojejunostomy was performed. Histopathologically, a submucosal adenocarcinoma arising from HP tissue was found. Although rare, gastric HP should be always kept in mind in patients presenting with pyloric mass causing gastric outlet obstruction.

**Keywords**

Gastric Cancer, Heterotopic Pancreas, Obstruction

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## Introduction

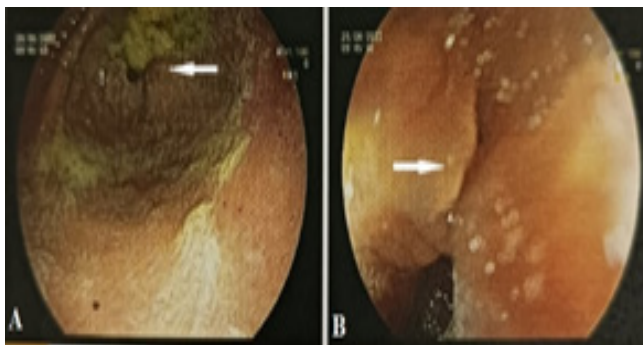
Heterotopic pancreas (HP), described as the abnormal location of pancreatic tissue without anatomical or vascular connection to the original pancreas, is usually found incidentally during the endoscopic examination, surgical procedure or autopsy. It is a rare developmental entity with an estimated incidence of up to 1.2% on the basis of abdominal surgery [1]. HP is usually asymptomatic or has nonspecific symptomatology such as abdominal pain and dyspepsia. However, the clinical presentation mainly depends on the site, size, and histological nature of the ectopic tissue [2].

Although HP can be found anywhere in the body, the stomach is the most common location [3]. Most gastric cases have benign behavior, and approximately 20 well-documented malignant cases have been reported in the literature [4]. However, a very limited number of cases of malignant HP presenting with gastric outlet obstruction have been published to date [5-7]. In this paper, an extremely rare case of obstructive gastric adenocarcinoma arising from HP was reported and discussed in light of the literature.

## Case Report

A 69-year-old man presented to our hospital with epigastric distension, nausea, vomiting, and 4 kg weight loss for two months. The patient had an unremarkable medical history. Upper gastrointestinal endoscopy revealed a dilated stomach filled with food debris and a prepyloric submucosal wall thickening that caused a complete gastric outlet obstruction (Figure 1A, B). No significant ulceration or inflammation was observed on the mucosal surface, and a biopsy was taken from the mucosa in the narrowed area. The biopsy result was reported as chronic active gastritis without evidence of malignancy. Abdominal computed tomography (CT) demonstrated dilated gastric lumen with circumferential wall thickening in the pylorus (Figure 2). There was also no distant or lymphatic metastasis. Laboratory tests, including CA19-9 were within normal limits.

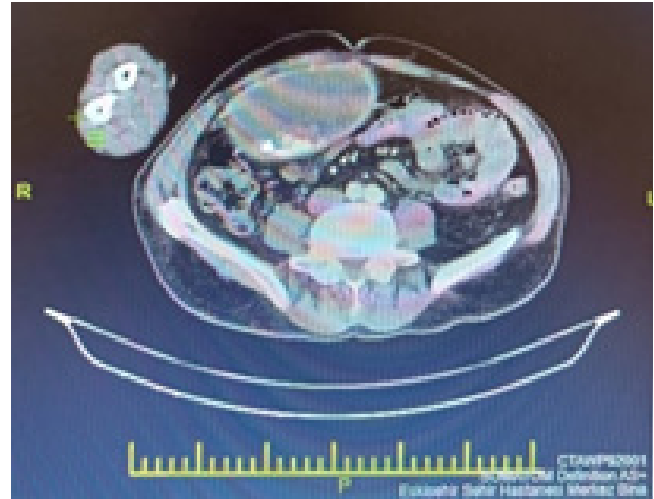
A surgical approach was planned for the patient with the diagnosis of pyloric stenosis. At laparotomy, the stomach was highly dilated and edematous, and a firm mass obstructing the lumen was palpated in the pyloric region. No additional pathology was found. A subtotal gastrectomy with gastrojejunostomy was performed for the pyloric tumor. In the histopathological examination, a well differentiated adenocarcinoma of 3.5×2.5×2 cm was found. Microscopically,



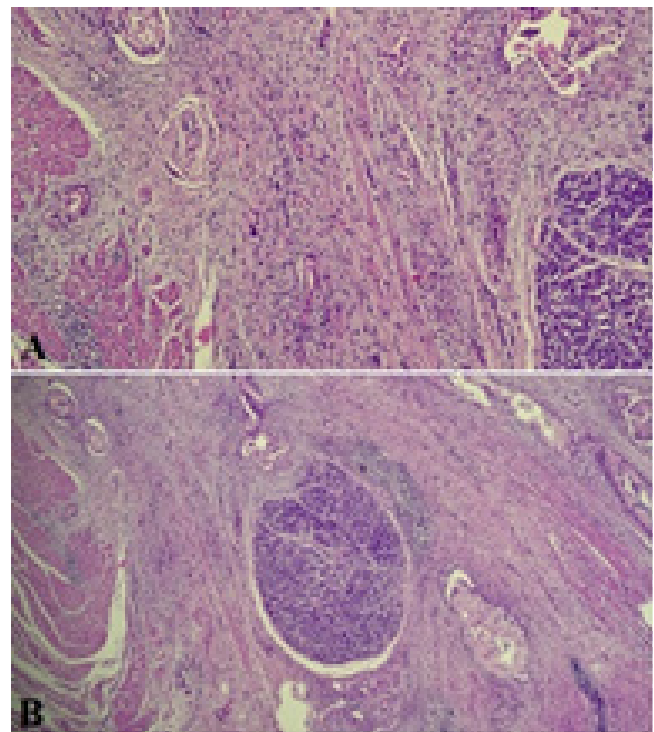
**Figure 1.** (A) stenotic pylorus (B) Pyloric deformation that completely obstructs the passage to the duodenum (submucosal wall thickening).

the tumor was located at the submucosa, and the mucosa over the tumor was intact. However, the tumor cells were found to be positive for CK7, CK 19, CA19-9, CEA, and MUC1, and there were multiple focal pancreatic tissues within the tumor, indicating the HP. Non-neoplastic heterotopic pancreatic tissue also showed ducts and acini. As a result, the tumor was reported as an adenocarcinoma arising from the HP (Figure 3A, B).

The patient was uneventfully discharged on the postoperative 10<sup>th</sup> day, and was referred to the medical oncology unit for the continuation of oncological treatment. Informed consent was obtained from the patient.



**Figure 2.** Tomographic appearance of the dilated gastric lumen with circumferential wall thickening in the pylorus.



**Figure 3.** (A) Ectopic pancreas (right bottom) and ductal adenocarcinoma with irregular glands, desmoplasia and cytologic atypia (Hematoxylin and eosin staining, x10), (B) Ductal adenocarcinoma arising from ectopic pancreas (Hematoxylin and eosin staining, x4).

## Discussion

Although various hypotheses have been proposed about the pathogenesis of HP, the exact cause is still unclear. The most common theory is the accumulation of pancreatic tissue fragments in ectopic areas during the embryological rotation of the foregut. Therefore, it is not surprising that the most common site of the presentation is within 5 cm of the pylorus, which is anatomically closest to the original pancreas [8]. Similarly, a prepyloric tumor causing a gastric outlet syndrome was detected in our patient.

Most of the cases of HP are clinically silent and found incidentally during endoscopic or surgical interventions performed for other reasons. Complicated situations such as bleeding and obstruction can develop in a small number of patients with HP [7]. In a recent study, abdominal pain was determined as the most common symptom, while only 9% of the patients with HP developed gastric outlet obstruction [3]. Malignant transformation, on the other hand, is an extremely rare condition with a reported incidence of under 1% in patients with HP. To the best of our knowledge, there are only two reports of gastric adenocarcinoma arising from a HP and causing gastric outlet obstruction [4, 5].

Due to the limited number of cases reported in the literature, there are no specific age and gender predilection for malignant HP. However, two cases have been reported with both malignant transformation and gastric outlet obstruction: a 75-year-old man and a 44-year-old woman. Our case was a 69-year-old man.

The preoperative diagnosis of HP is quite difficult due to the nonspecific clinical and radiological features. A well-circumscribed submucosal lesion with central umbilication was reported as the typical endoscopic finding of gastric HP [6]. However, this finding is found in less than half of the patients, and the endoscopic appearance is similar to other types of neoplasia including submucosal carcinomas, gastrointestinal stromal tumors, neuroendocrine tumors, and leiomyomas [7]. As a result, the definitive diagnosis is almost always made by histopathological evaluation of the lesion. Since HP is a submucosal lesion, multiple and deep biopsies during the endoscopic examination are of great importance for the correct diagnosis.

Although the treatment of incidentally detected benign and asymptomatic lesions is controversial, endoscopic submucosal dissections in appropriate cases or limited surgical resections from localized resection to antrectomy are recommended for symptomatic patients. In malignant cases, depending on the location of the tumor, radical oncological operations, including lymphatic dissection are essential [5, 6]. The present case had a prepyloric obstructive tumor, thus radical subtotal gastrectomy together with D2 lymphatic dissection was performed.

## Conclusion

Although rare, gastric HP should be always kept in mind in patients presenting with pyloric mass causing gastric outlet obstruction. Malignant cases should be treated in accordance with the oncological principles of gastric cancer.

## Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some

of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

## Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

## Conflict of interest

None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.

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